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Sialo-cutaneous fistula caused by submandibular gland sialolith



KEYWORDS

Sialo-cutaneous fistula;
Sialolith;
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Sialoliths commonly occur in the duct or parenchyma of the submandibular gland (SMG), and the fistulae formation to other compartments is rarely caused by sialadenitis and sialolithiasis.¹ Although sialo-oral fistulae are rarely caused by the obstruction of the Wharton's duct and inflammatory reaction,^{1,2} sialo-cutaneous fistulae associated with SMG sialoliths are rarer.^{1–5} Sialo-cutaneous fistulae generally present with serous/mucoid/mucopurulent discharge from the opening, and the fistulae associated with sialolith mostly present with obstructive or colicky complaints of pain/tenderness/swelling in the submandibular region.⁴ We reported a sialo-cutaneous fistula caused by SMG sialolith.

A 49-year-old woman had recurrent pain and swelling of the left SMG. The mucosa of the left oral floor was intact (Fig. 1A). For the cervical subcutaneous fistula (Fig. 1B and C), the patient was referred to our hospital. There was no history of submandibular trauma. Computed tomography (CT) showed a sialolith (7 × 6 × 6 mm) in the duct of the left SMG and sialo-cutaneous fistula (Fig. 1D–F). Magnetic resonance imaging (MRI) revealed saliva in the sialo-cutaneous fistula as well as the duct and parenchyma of the SMG (Fig. 1G–I). Clinical and radiological diagnosis was SMG sialolith and sialo-cutaneous fistula. Because the patient hoped for SMG preservation, the fistula excision and intraoral removal of the sialolith with sialodochoplasty under general anesthesia. A spindle-shaped skin incision around the fistula was made, and the fistula was dissected and followed to the SMG (Fig. 1J and K). The fistula was ligated and cut, and the SMG was preserved. After skin

suture, intraoral sialolith removal with sialodochoplasty was performed (Fig. 1L). The postoperative course was uneventful without recurrence of the fistula.

Sialo-cutaneous fistulae caused by SMG sialoliths are very rare.^{1–5} Wakoh et al.¹ reported a rare case of sialo-cutaneous fistula containing a large, ectopic sialolith in subcutaneous tissue, although sialoliths are commonly located in the duct or parenchyma of the SMG in cases with sialo-cutaneous fistula.

In a recent radiological review,¹ SMG sialo-fistulae related to sialolithiasis was classified into four types from the drainage routes: sialo-oral fistula, sialo-cutaneous fistula, sialo-pharyngeal fistula, and sialo-oro-cutaneous fistula. Sialo-cutaneous fistulae were found in 10 cases (including a case of sialo-oro-cutaneous fistula), while only 4 cases identified the fistulous tract on radiological imaging such as CT and fistulography.¹ In the present case, sialo-cutaneous fistula was identified by CT and MRI.

A systematic review of SMG fistulae showed 13 of 18 cases (72.2 %) had sialo-cutaneous fistulae.⁴ Five (including a case of sialo-oro-cutaneous fistula) of 18 cases (27.8 %) had sialoliths as the cause. Three of 5 cases with SMG sialoliths underwent SMG removal, and one underwent sialendoscopic removal of sialolith (5 × 4 mm) and sialo-cutaneous fistula excision. The remaining one with sialo-oro-cutaneous fistula underwent removal of sialolith and sialo-cutaneous fistula.³ Because the present case had the sialolith in the Wharton's duct and sialo-cutaneous fistula, sialo-cutaneous fistula excision and intraoral sialolith

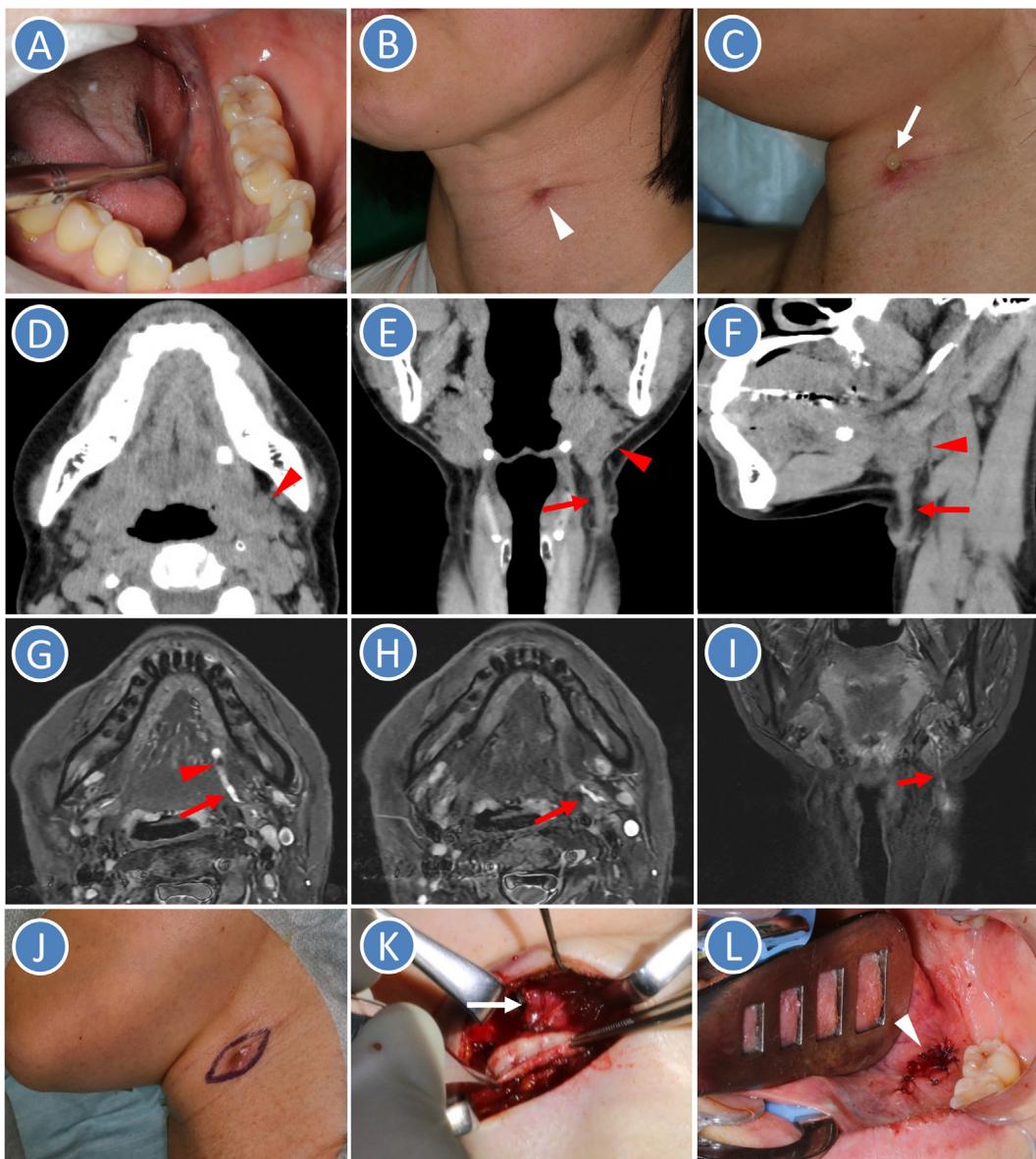


Figure 1 Intraoral and extraoral photographs and radiological images. (A) Intraoral photograph. (B,C) Extraoral photographs. Arrow indicates mucous saliva. Arrowhead indicates fistula. (D–F) Computed tomography. Arrow indicates fistula. Arrowhead indicates submandibular gland. (G–I) Magnetic resonance imaging. Arrow indicates saliva. Arrowhead indicates sialolith. (J–L) Intraoperative photographs. Arrow indicates fistula. Arrowhead indicates orifice after sialodochoplasty.

removal with sialodochoplasty were performed for preservation of the SMG. Depending on the location and size of sialoliths, SMG preserving surgery with sialo-cutaneous fistula excision can provide a good prognosis.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

Acknowledgments

None.

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